

Mini Review

Impact of cleft lip and/or cleft palate in sleep-breathing disorders. An updated mini-review

Ana Boquete Castro*, Cortes-Mejia JM

Postgraduate student of the Master in Dental Sleep Medicine, UCAM, Madrid, Spain

*Corresponding author: Ana Boquete Castro, Postgraduate student of the Master in Dental Sleep Medicine, UCAM, Madrid, Spain

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Introduction

Cleft lip with or without cleft palate is one of the most common congenital malformations, with a prevalence around 1 case each 100 births [1]. The main cause is cleft-palate is the appearance of de-novo mutations [4]. Impellizzeri et al. carried out a retrospective study and observed a female prevalence for cleft palate and a male predominance for cleft lip. 70% of the cases course without other associated pathologies [11]. These congenital defects usually affect normal deglutition and speech of patients [12].

Children with cleft lip or palate are known to have airway complications [1]. In children that present craniofacial syndromes, the first cause for hospitalization is the obstruction of the airway, especially during neonatal period [4]. There are more than 200 syndromes that course with cleft palate, including sometimes also cleft lip, and which usually have narrow upper airways and mandible retrognathia which favors the obstruction at the upper airway. Moreover, is common the presence of nasal deformities that worsen the obstruction of the airways [4].

Cleft palate appears as a consequence after an inadequate fusion between medial and lateral nasal processes with the upper maxilla. Children with cleft palate have functional and structural changes in their upper airways, thus increasing the risk of suffering from sleep-breathing disorders [9]. There is a smaller dimension of the oropharyngeal height and the airway volume is reduced when compared with non-cleft individuals [1]. According to the literature, maxilla-mandibular advancement can enlarge the pharyngeal airway [3].

It does not exist an established definition of

Abstract

Cleft palate is one of the most common congenital malformations. There are more than 200 syndromes that course with cleft palate, including sometimes also cleft lip. Children with cleft lip or palate are known to have airway complications and more probability of suffering from obstructive sleep apnea (OSA). To determine the severity of OSA in children is crucial, because it can lead to several problems such as: altered growth or cognitive defects, among others. Treatment options for these patients implicate a multidisciplinary approach in order to obtain an increase of the airway volume.

Keywords: cleft lip, cleft palate, obstructive sleep apnea.

breathing-sleep disorders in children [9]. Obstructive sleep apnea (OSA is the most commonly described sleep-breathing disorder in children with craniofacial syndromes [11]. The presence of sleep-breathing disorder in children has negative consequences and is associated with several comorbidities such as growing disturbances, cognitive compromise and poor life-quality because of the difficult feeding [9].

Children with craniofacial syndromes have more probability of suffering from OSA [6]. As a result, these patients must be carefully evaluated and diagnosed in order to choose the best treatment option for each individualized case [4].

In addition, children with cleft anomalies have more psychological issues. The attitude of parents is a key factor in the child's perception of their impairment [10].

Methodology

An updated literature review was carried out via Medline to analyze the current status of cleft lip and cleft palate and its relationship with breathing disorders during sleeping. Electronic bibliographic search was carried out in November 2019, among using the MeSH terms: "sleep apnea, obstructive", "cleft lip" and "cleft palate". Articles published more than 5 years ago and those published in a language different from English were excluded.

Discussion

Clinical findings are variable in children with cleft lip or palate. Dental findings in individuals with cleft lip or palate usually include maxillary retrusion and posterior crossbite [1]. According to Chang et al. up to 60% of cleft patients will require orthognathic surgery.

In addition, cleft lip and cleft palate are relatively common pathologies that can occur with other concomitant pathologies such as congenital heart disease [5]. Thus, children with cleft lip or palate need a multidisciplinary evaluation and treatment by plastic surgeon, dentist and ENT among other specialists [4].

Diagnosis

To determine the severity of OSA in children is crucial. Analysis of clinical signs such as snoring or pulsioximetry are not enough. Polysomnography (PSG) is the gold standard for diagnosis in newborn [4] because it gives an objective

analysis of breathing parameters during sleeping, and quantification of the obstruction of the upper airway [7].

Until some years ago, lateral-crane radiography was used to set the characteristics of the upper airway; however, this technique is limited and can produce superpositions, magnification, etc. Nowadays, cone beam tomography is considered the best image tool for diagnosis because it proportionates good quality images with an accurate determination of the anatomical structures without superpositions [5].

Smith et al. followed-up 52 children from born to three years of age. A PSG was carried out in every children and neurocognitive development (speech, motor attitude, etc.). The authors concluded that children with sleep-breathing disorders give rise to intermittent hypoxia episodes, neuronal alterations and growth alterations.

In addition to PSG, physical examination is important in order to determine the degree of micrognathia, facial hypoplasia, glossoptosis or other disturbances [4].

Treatment

Physical appearance plays an important role in the development of self-beliefs [10]. Usually, children affected by cleft lip and/or palate have a diminished social acceptance. Moreover, those children usually have language disorders that can be accompanied by hearing loss or facial disfigurement [12]. Surgery is the ideal option for solving most of the problems associated to cleft lip and palate [10] [8]. reported a case of complete cleft lip and alveolus with presence of severe nasal deformity. The case was treated with an acrylic presurgical nasoalveolar molding (NAM) appliance that was adjusted weekly to gradually lengthen the columella. The authors concluded that after 3 months of use, lips were approximated and columella was lengthened, favoring the result of the subsequent lip surgery thus reducing the number of future surgeries needed [8].

Moreover, the treatment of the upper airway will result in a benefit because feeding will get better [11].

Usually, first treatment option in children suffering OSA and with cleft palate is to carry out a tonsillectomy [11]. However, it exists the risk of a velopharyngeal insufficiency, so a detailed examination must be realized before [6].

Some authors have described the use of individualized acrylic removable devices that avoid the tongue entrance on the nasal cavity as a treatment of OSA in children. In this sense, some studies have analyzed the changes of measurements of the upper airway after nasoalveolar modelling. This technique is not only focused in correcting the alveolus, but also the lip and the nose during the first months of life and before lip surgery [2]. It leads to a better nasal aesthetics, repositioning of the filtrum and elongation of the columella; moreover, it reduces the extension of the palatal opening and expands upper maxilla, among other benefits for the patient [2].

Thakur & Kapoor reported a case of a 6 year-old female with unilateral cleft lip and palate. She presented a distance of 8mm between both nasoalveolar segments. Impressions were taken in order to obtain an acrylic NAM plate with a retentive button in the anterior region. The plate was activated by adding resin progressively. The authors obtained a reduction of the nasoalveolar cleft from 8 mm to 3 mm. Moreover, the authors obtained a lengthening of the columella and a improvement of the nasal symmetry prior to surgery [12].

Surgical procedures can also be carried out such as mandibular distraction. Results vary among patients and it is not possible to find a technique that stands out the others [11]. Some children continue presenting OSA after those interventions. Traqueostomy is the unique technique that has demonstrated to be the definite method to compensate the breathing deficit but is a high-complexity procedure, with other risks and associated comorbidities [11]. The scientific literature establishes the necessity of a multidisciplinary collaboration for an adequate management of these patients (plastic surgeon, maxillofacial surgeon, dentist, ENT, speech therapist, etc.) [11].

Chang et al. treated 18 consecutive patients with conventional orthognathic surgery. The authors concluded that, 6 months after surgery, patients have no improvement regarding AHI (apnoea/hypopnoea index) or lowest oxygen saturation (LSAT) despite velopharyngeal airway was increased after orthognathic surgery to advance the maxilla.

Some authors suggest that the obstruction of the airway will get better spontaneously because of the growth of the children, being able to solve up to 50% of cases without any intervention [4]. Alrejaye et al. reported a statistically significant increase in the oropharyngeal airway volume by measuring airway dimensions in CBCT scans; the authors obtained a statistically significant increase in pharyngeal airway volume after a orthodontic phase for maxillary expansion and protraction.

Hitherto, after treating these children, complete resolution of OSA is low and difficult to predict [11]. It is necessary to continue the research related to sleep-breathing disorders in children with cleft palate to understand its implications and to develop long-term successful treatments [9].

Conclusions

Cleft palate with or without cleft lip is the most common congenital malformation in the craniofacial region. It is very important to treat this individual as soon as possible in order to avoid airway complications and other disabilities related to feeding or speech.

PNAM has proved to be an efficient adjuvant therapy for reducing hard and soft tissue deformities in infants with cleft and lip palate before plastic surgery.

References

1. Alrejaye N, Gao J, Hatcher D, Oberoi S. Effect of maxillary expansion and protraction on the oropharyngeal airway in individuals with non-syndromic cleft palate with or without cleft lip. *PLoS One*. 2019; 14[7]: e0213328.
2. Astani SA, Yilmaz HN, Nevzatoglu S, Demirkaya AA, Acar ZA. Evaluation of Airway Volume in Cleft Lip and Palate Following Nasoalveolar Molding. *J Craniofac Surg*. 2018; 29:2143-2147.
3. Chang CS, Wallace CG, Hsiao YC, Hsieh YJ, Wang YC, and Chen NH et al. Airway Changes after Cleft Orthognathic Surgery Evaluated by Three-Dimensional Computed Tomography and Overnight Polysomnographic Study. *Sci Rep*. 2017;7[1]:12260.
4. Cielo CM, Montalva FM Taylor JA. Craniofacial disorders associated with airway obstruction in the neonate. *Semin Fetal Neonatal Med*. 2016; 21[4]: 254-262.
5. Impellizzeri A, Giannantoni I, Polimeni A, Barbato E, Galluccio G. Epidemiological characteristic of Orofacial clefts and

its associated congenital anomalies: retrospective study. *BMC Oral Health*. 2019; 23; 19[1]:290.

6. MacLean JE, Fitzsimons D, Fitzgerald D, Waters K. Comparison of clinical symptoms and severity of sleep-disordered breathing in children with and without cleft lip and/or palate. *Cleft Palate Craniofac J*. 2017; 54[5]:523-529.

7. MacLean JE. Sleep frequently asked questions: Question 1: What abnormalities do babies with cleft lip and/or palate have on polysomnography? *Paediatric Resp Rev*. 2018; 27:44-47.

8. Premkumar S, Kunthavai CR, Tovani-Palone MR. Clinical application of customized presurgical nasoalveolar molding for the treatment of unilateral complete cleft lip and alveolus: case report. *Electron J Gen Med*. 2018;15[5]:em76.

9. Smith CB, Walker K, Badawi N, Waters, KA. Impact of Sleep and Breathing in Infancy on Outcomes at Three Years of Age for Children with Cleft Lip and/or Palate. *SLEEP* 2014; 37[5]:919-925.

10. Sousa AD, Devare S, Ghanshani J. Psychological issues in cleft lip and cleft palate. *J Indian Assoc Pediatr Surg*. 2009; 14[2]:55-58.

11. Tan HL, Kheirandish-Gozal L, Abel F, Gozal D. Craniofacial Syndromes and Sleep-Related Breathing Disorders. *Sleep Med Rev*. 2016; 27:74-88.

12. Thakur S and Kapoor AK. Treatment of unilateral complete cleft lip and palate using pre-surgical nasoalveolar molding (PNAM) therapy: a case report. *Int J Res Health Allied Sci*. 2017; 3[1]:23-25.